# Colonic Duplication Cyst with Gastric and Pancreatic Heterotopia: A Case Report

*Mwenyemali Tony*[1], *Odundo Sam*[2], *Opondo Dedan*[1], *Mbayah Etabale*[3], *Awuonda Bernard*[3]

[1]Aga Khan Hospital Kisumu, Surgery, Kisumu, Nyanza, Kenya

[2]Nyanza Provincial General Hospital, Surgery, Kisumu, Kenya

[3]Maseno University Kisumu, Nyanza, Kenya

***Correspondence to:*** Mwenyemali Tony; email: bmwenyemali@gmail.com

Received: 15 Jan 2022; Revised: 18 Nov 2022; Accepted: 19 Nov 2022; Available online: 24 Nov 2022

**Summary**

Colonic duplication is an uncommon congenital anomaly within the alimentary tract which may be missed on clinical examination. Clinical symptoms are generally related to the involved site, size of duplication, or the associated ectopic mucosa. This is a case report of a 7-month-old female patient admitted in septic and hypovolemic shock, with severe anemia. Ultrasonography and computed tomography (CT) scans showed a left flank mass suggesting intussusception. The patient was first stabilized with intravenous fluids, intravenous antibiotics, oxygen, and blood transfusion. At laparotomy, the findings were a left paracolic cystic mass and an adjacent, complex retroperitoneal hemorrhagic mass. The histopathology report was consistent with a colonic duplication cyst with ectopic gastric and pancreatic mucosa. The patient improved after surgery and was discharged for routine outpatient follow-up. Colonic duplication should be included in the differential diagnoses in children presenting with palpable abdominal masses. Complete surgical removal of the symptomatic duplication is the treatment of choice.

**Keywords**: Duplication cyst, Resuscitation, Laparotomy, Histopathology, Case report

Ann Afr Surg. 2023; 20(2): 63-68
**DOI**: http://dx.doi.org/10.4314/aas.v20i2.5

**Conflict of interest: None**
**Funding:** None


## Introduction

Alimentary tract duplication (ATD) represents 0.2% of all pediatric congenital malformations with a reported incidence of 1:4,500 births. They can present anywhere in the gastrointestinal tract (GIT) from the mouth to the anus and have no familial or racial predilections. Duplicated colon is either cystic or tubular; it is a rather unusual clinical entity that remains asymptomatic and undiagnosed in most cases (1).

GIT duplication cysts are a rare congenital gastrointestinal malformation in young patients and adults consisting of foregut, small bowel, and large bowel duplication cysts. Colonic duplication cysts represent 6.8% of gastrointestinal duplications and 33% may contain heterotopic gastric mucosa (2).

The duplication may be on the mesenteric or anti-mesenteric side. It may share the same vascular supply, mesentery, and lumen with the bowel.

Most duplications are detected in children, antenatally or within the first 2 years of life, and fewer than 30% of all duplications are diagnosed in adults (3).

There are two types of duplication cysts: (i) cystic duplications which are more common (80%) and are spherical in shape with no communication with the bowel lumen; and (ii) tubular duplications that may communicate with the normal intestine at one or several points along the common wall and usually extend above and below the diaphragm (2). Excision of the duplication is recommended due to reported risk of malignancy (1). Although it is rare, colonic duplication needs to be listed among important differential diagnoses in children with recurrent abdominal pains and vomiting.

**Case presentation**

A 7-month-old female patient weighing 6.3 kg was brought to the emergency department (ED) with a history of recurrent vomiting and diarrhea for 2 weeks with an accompanying 4-day history of fever and abdominal pain. On examination, she was lethargic, had cold extremities, sunken eye balls, reduced skin turgor >3 seconds, weak peripheral pulses, prolonged capillary refill >3 seconds, altered level of consciousness (AVPU = P), severely pale, and had tachycardia. Abdominal examination revealed tenderness and a palpable left flank mass which was thought to be intussusception. Attempts at peripheral, intraosseous, and central vein catheterization failed, leading to emergency venous cut-down. Blood samples were collected for complete blood count, urea electrolytes and creatinine, blood grouping and cross matching, serum procalcitonin, liver function tests, and hemoglobin (HB) electrophoresis. A stool sample was collected for microscopy, occult blood, and rotavirus/adenovirus antigen tests. A bedside abdominal pelvic ultrasonography (US) done showed minimal ascites and a 2.8 × 2.5 cm2 circular bowel structure in the left lumbar region with a thickened wall and fluid accumulation overshadowing the spleen. Blood works revealed leukocytosis of 36 × 109/L, microcytic hypochromic anemia of 3 g/dL, elevated procalcitonin of 181 ng/mL, metabolic acidosis, deranged liver function tests, elevated serum creatinine, elevated lipase, and reduced serum amylase and hemoglobin (HB) electrophoresis showed normal hemoglobin (HbAA). Stool tests were negative for ova and cysts, occult blood, adenovirus, and rotavirus.

The patient was resuscitated at the ED with intravenous fluids (Hartman's solution 20 mL/kg bolus administered twice 20 min apart), oxygen (via mask at 5L/min) and blood transfusion (packed red cells at 10mL/kg in 3 hours with an additional two units during the hospital stay. She was also started on IV antibiotics (Ceftriaxone at 50 mg/kg bolus and maintained every 12 hours for 7 days) and admitted to the High Dependency Unit (HDU). Computed tomography (CT) scan of the abdomen showed left lumbar mass suggestive of intussusception (Figures 1–3).

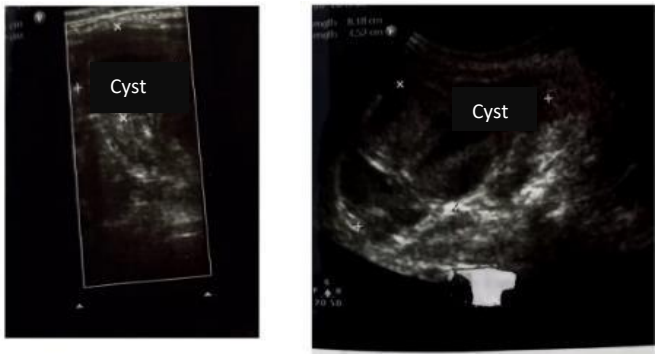



Figure 1. Abdominal pelvic US showing minimal ascites, 2.8 × 2.5 cm2 size circular bowel structure with a thickened wall and fluid accumulation.

The patient was prepared for emergency laparotomy after resuscitation and stabilization.

During laparotomy, a 7 × 3 × 3 cm3 soft, gray, cystic mass was found between the stomach and the spleen. It was attached to the pancreatic tail and was partially adherent to the distal transverse and descending colon. The cystic tumor had a thick wall, did not communicate with the gastrointestinal lumen, and was found on the anti-mesenteric side of the colon. Its blood supply was from the splenic vessels.

Another mass measuring 8 × 4 × 3 cm3 was found in the retroperitoneum behind the descending colon and sigmoid colon. It displaced the left kidney, aorta, and left colon medially and had no gastrointestinal communication. It was a dark red mass with a yellow

portion toward its upper pole with a complex friable and hemorrhagic structure. Its blood supply was from the retroperitoneum. The two tumors were separate.

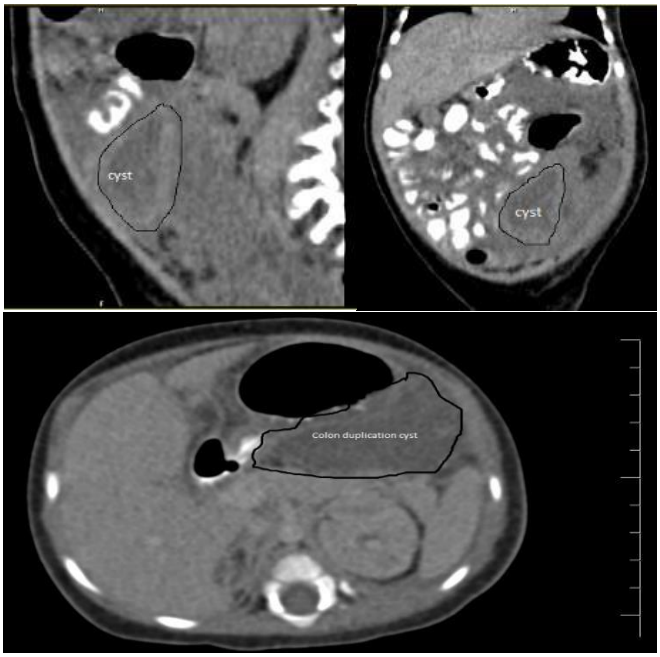



Figure 2. Axial and coronal abdominal CT scan showing cystic mass.

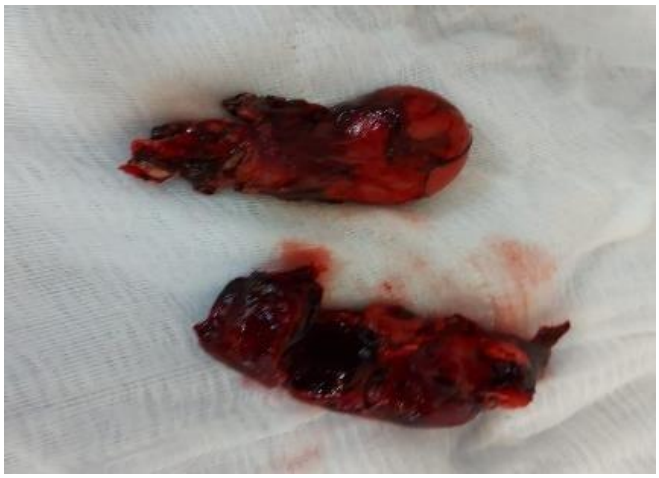
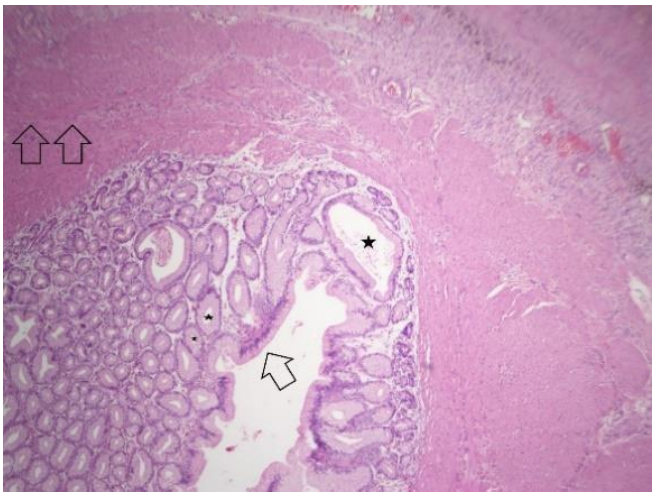
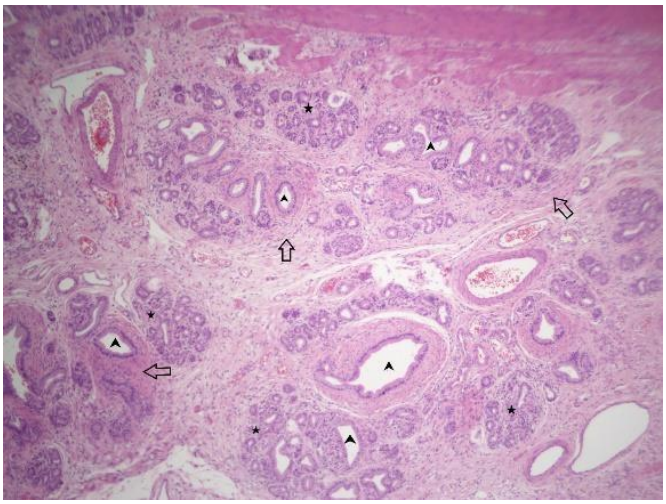


Figure 3. Excised cystic mass (upper mass) and complex mass (lower mass).

A retroperitoneal hematoma extending from the left colonic gutter toward the liver and right colon was also present. These tumors were completely excised and a peripancreatic closed suction drain was inserted in the lesser sac.



(A) Histopathology of the resected specimen showed a gut segment whose mucosa is composed of gastric foveolar epithelium (arrow) and gastric pits (stars). The muscularis propria (double arrow) is also represented. These findings are in keeping with the ectopic gastric tissue (gastric heterotopia) in a colonic duplication cyst. Other ectopic tissues that may be encountered in a colonic duplication cyst include pancreatic, thyroid, and bronchial tissues.



(B) Within the muscularis propria and serosa of the resected specimen are acinar lobules (star) and their corresponding ductules (arrowheads). The acini are atrophic with mild chronic inflammatory cell infiltration and fibrosis (arrows) noted in the intralobular and interlobular stroma. The ducts and ductules are dilated. These features are consistent with chronically inflamed ectopic pancreatic tissue.

Histopathology of the cystic mass showed a structure containing a mucosa, submucosa, muscularis propria, and serosa. The mucosa was of gastric type with

evidence of ulceration. Some acini and ducts compatible with pancreatic tissue were noted in the outer muscularis propria and serosa. Recent and old hemorrhagic material mixed with inflammatory infiltrate and granulation tissues were present within the lumen and in the cyst wall.

Histopathology for the retrocolic hemorrhagic mass showed no recognizable normal tissue. The tissue had undergone complete hemorrhagic infarction.

The patient had an uneventful post-operative course and was discharged after 5 days. The pancreatic drain was removed prior to discharge.

**Discussion**

Enteric duplication cysts are rare congenital anomalies that can be found at all levels of the alimentary tract. A majority of these cysts are symptomatic within the first 2 years of life with varied clinical presentations and require both radiological and histopathological evaluation for diagnosis (4).

As per Ladd, the term "enteric duplication cyst" will be applied if a congenital lesion meets the following criteria: (i) it has a coat of smooth muscle; (ii) it has a GI-type epithelial lining; and (iii) it has intimate anatomical location with some part of GIT (5).

Although 80% of cases with ATDs are detected in the first 2 years of life, duplications can remain undetected until an advanced age. We should keep in mind the potential for malignant change in patients whose duplications are detected in old age. Although the etiology is currently unknown, duplications are believed to occur between the 4th and 8th weeks of embryological development (6).

The ileum is the most common site for a duplication cyst (30–35%), while the colon is the least common site (7–20%). The transverse colon is the most common site for colonic duplication. These lesions occur so infrequently that they are often not suspected until encountered intraoperatively (1).

Several theories have been proposed for the development of ATDs but none can solely explain all the locations where this anomaly may occur. An intriguing theory supposes a twinning process involving the hindgut before the primitive intestinal tube has been entirely formed. When twinning occurs early, all the structures derived from hindgut (ileum, colon, rectum, urinary bladder, and urethra) will be doubled. An alternative theory is that the gut begins as a solid structure of crowded epithelial cells. As these cells are rearranged and absorbed, vacuoles appear that eventually coalesce to form the bowel lumen. On occasion, vacuoles may coalesce parallel to the normal segment of the gut forming a duplication (1).

The most common clinical presentation is abdominal pain with signs and symptoms resulting from intestinal obstruction. Presentations can also include gastrointestinal bleeding, intussusception, or as an incidental finding (Table 1). This may depend on the size, location, type, and presence of ectopic mucosa (6). In our case, this was an incidental finding during exploratory laparotomy.

Table 1. Case reports on colonic duplication cysts

| Sample no. | Author | Year | Age | Part of gut involved | Symptoms |
|---|---|---|---|---|---|
| **1** | Takrouney MH, et al. (1) | 2020 | 18 Months | Colon | Grayish substance from the genitalia |
| **2** | Sharma S, et al. (3) | 2015 | 3 days–10 years | Small and large bowel | Abdominal pain, distennsion, vomiting |
| **3** | Tiwari C, et al. (5) | 2017 | 4 years | Small and large bowel, omentum, mesentry | Abdominal pain |
| **4** | Kim SH, et al. (6) | 2020 | 1 day more than 60 months | Foregut, midgut, and hindgut | Features of intestinal obstruction |
| **5** | Garg R, et al. (7) | 2018 | 42 years | Colon | Features of intestinal obstruction |
| **6** | Bhadoo D, et al. (8) | 2015 | 3 years | Colon | Constipation and rectal bleeding |
| **7** | Cheng KC, et al. (9) | 2019 | 29 years | Colon | Chronic constipation, vomiting, and fever |

Duplication cysts can also have ectopic tissue of two origins, most commonly heterotopic gastric mucosa,

which can ulcerate leading to bleeding, and less commonly pancreatic tissue (7).

In our case, the histopathology detected ectopic gastric mucosa and ectopic pancreatic tissue disseminated in the muscularis propria and serosa of the duplication cyst.

Gastrointestinal bleeding in bowel duplications can result from angiodysplasia or ectopic gastric mucosa leading to peptic ulceration, bleeding, and perforation with peritonitis (8).

Enteric duplications in association with the pancreas are especially uncommon. Pancreatic duplication cysts may present in a variety of ways but are most commonly associated with abdominal pain. Devastating presentations with peritonitis and severe pancreatitis have been reported (6, 8).

The ATD lesion, especially the cystic type with non-communication to the adjacent normal bowel, generally tends to increase in size over time and cause related symptoms (6).

The management of asymptomatic colonic duplication is not well established. Some authors suggest that surgical resection should be reserved for symptomatic cases, though others suggest resection once the diagnosis is made to prevent potential complications, including malignant change (9). The most common malignancy is adenocarcinoma, followed by squamous cell carcinoma and carcinoid tumor with mean age at diagnosis being 48.8 years (range of 23–72 years). The preoperative diagnosis of malignant transformation is difficult, but should be considered when a solid component is found within a duplication accompanied by high serum levels of CA19-9 or carcinoembryonic antigen (CEA) (10).

Here, we report a very rare case of cystic colonic duplication without any other congenital defect in a child, presenting with a palpable abdominal mass and hypovolemic shock with severe anemia.

The surgical specimen showed a cystic duplication extending craniocaudally from the tail of the pancreas to the junction between the descending and sigmoid colon with attachment at the splenic hilum. No communication was noted between the native colon and the duplicated lumen. Histopathology showed a colonic duplication cyst with gastric and pancreatic heterotopia.

The second mass, completely separate from the first, was in the retroperitoneum behind the descending colon and appeared complex and hemorrhagic. Microscopic examination revealed a tissue with complete hemorrhagic infarction. There is a probability that this mass was a second colonic cystic duplication that developed an ulceration, bleeding, and then perforation. And this would explain the hypovolemic shock, severe anemia, and sepsis that the patient presented with on admission.

Although duplication cysts are rare and of varied clinical presentation, hemorrhage from the cysts can lead to severe anemia with shock. This requires a high index of suspicion in children with palpable abdominal masses and radiological findings suggestive of intussusception. Most authors recommend that once the diagnosis is made, an elective surgical procedure should be performed to avoid complications and that the procedure should be performed in a stable patient, but few think that only symptomatic duplications should be surgically treated (9).

In our case, the patient was critically ill and an emergency surgery was warranted after resuscitation. The recommended surgical procedure is excision of the duplication (9). Laparoscopic approach can be utilized for select cases.

## Conclusion

Colonic duplications are rare but should be kept in mind in the differential diagnoses of an acute abdomen in children. It can present in different clinical forms, and if left untreated can cause significant morbidity and mortality through obstruction, ulceration, bleeding, perforation, and malignant transformation.

## Ethical consideration

Informed consent was acquired from the patient for publication of the case report.

## Author contributions

All other authors contributed equally in the conceptualization and writing of the first draft to reviewing and editing the original draft.

## References


1. Takrouney MH, Abdelmohsen SM, Osman MA, et al. Tubular colonic duplications. J Pediatr Surg Case Reports. 2020; 61: 101499.
2. Liu R, Adler DG. Duplication cysts: diagnosis, management, and the role of endoscopic ultrasound. Endosc Ultrasound. 2014; 3: 152–60.
3. Sharma S, Yadav AK, Mandal AK, et al. Enteric duplication cysts in children: a clinicopathological dilemma. J Clin Diagnostic Res. 2015; 9: EC08-EC11.
4. Anand S, Aleem A. Duplication cyst. StatPearls . NCBI Bookshelf, 2021.
5. Tiwari C, Shah H, Waghmare M, et al. Cysts of gastrointestinal origin in children: varied presentation. Pediatr Gastroenterol Hepatol Nutr. 2017; 20: 94–9.
6. Kim SH, Cho YH, Kim HY. Alimentary tract duplication in pediatric patients: its distinct clinical features and managements. Pediatr Gastroenterol Hepatol Nutr. 2020; 23: 423–9.
7. Garg R, Saravolatz LD, Barawi M. Endoscopic treatment of colonic duplication cyst: a case report and review of the literature. Case Rep Gastrointest Med. 2018; 2018: 1–5.
8. Bhadoo D, Chand K, Jana M, et al. Colonic duplication: treatment by limited division of common wall. J Indian Assoc Pediatr Surg. 2015; 20: 146–7.
9. Cheng KC, Ko SF, Lee KC. Colonic duplication presenting as a huge abdominal mass in an adult female. Int J Colorectal Dis. 2019; 34: 1995–8.
10. Rojas-Rojas MM, Mejiah M, Mora M, et al. Retroperitoneal mucinous neoplasm arising from colonic duplication cyst. J Gastrointest Cancer. 2019; 50: 583–5.